(International Print/Online Journal)

SJIF IMPACT FACTOR: 6.829
PUBMED-National Library of Medicine ID-101739732

ISSN (Print): 2209-2870
ISSN (Online): 2209-2862

International Journal of Medical Science and Current Research (IJMSCR)
Available online at: www.ijmscr.com
Volume 9, Issue 2 , Page No: 489-494
March-April 2026

IJMSCR

Adenamatoid Odontogenic Tumor In The Anterior Mandible: An Uncommon Pediatric Presentation

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Type of Publication: Original Research Paper
Conflicts of Interest: Nil

Abstract

Background: Adenomatoid odontogenic tumor (AOT) is a benign epithelial odontogenic tumor that predominantly affects adolescent females and most commonly involves the anterior maxilla, while its occurrence in the anterior mandible is rare.

Case Presentation: We report a case of an 11-year-old female presenting with a painless swelling in the anterior mandible. Radiographic examination revealed a well-defined unilocular radiolucency associated with an unerupted tooth 43. The lesion was surgically enucleated, and histopathological examination confirmed the diagnosis of adenomatoid odontogenic tumor (AOT).

Conclusion: Although AOT most commonly occurs in the anterior maxilla, it should also be considered in the differential diagnosis of anterior mandibular lesions in pediatric patients.
Early diagnosis and conservative surgical management ensure an excellent prognosis.

Keywords: Adenomatoid odontogenic tumor, Anterior mandible, odontogenic tumor

Introduction

Adenomatoid odontogenic tumor (AOT) is a rare, benign odontogenic epithelial tumor accounting for approximately 2–7% of all odontogenic tumors. It is characterized by a slow-growing and asymptomatic clinical behavior.1

Adenomatoid odontogenic tumor (AOT) has been known by several names over time, reflecting changes in understanding of the lesion. It was first reported by Steensland in 1905 and later termed "pseudo-adenoameloblastoma" by Dreibladt in 1907. In 1948, Stafne recognized it as a distinct entity, though it was still considered by some to be a variant of ameloblastoma. The term "adenomatoid odontogenic tumor" was introduced by Philipsen and Birn in 1969 and was officially adopted by the World Health Organization in 1971. Earlier names such as adenoameloblastoma, adamantinoma, and epithelioma adamantinum highlight the evolving concepts regarding its nature.2.

AOT usually occurs in the second decade, shows female predominance, and commonly involves the anterior maxilla, often with an impacted canine.

Radiographically, AOT appears as a unilocular radiolucency with a distinct radiopaque border, most commonly associated with an impacted tooth. Multiple small radiopaque foci or calcifications may

also be seen in some cases. Management of this odontogenic tumor involves surgical enucleation or curettage along with extraction of the associated impacted tooth.[3]

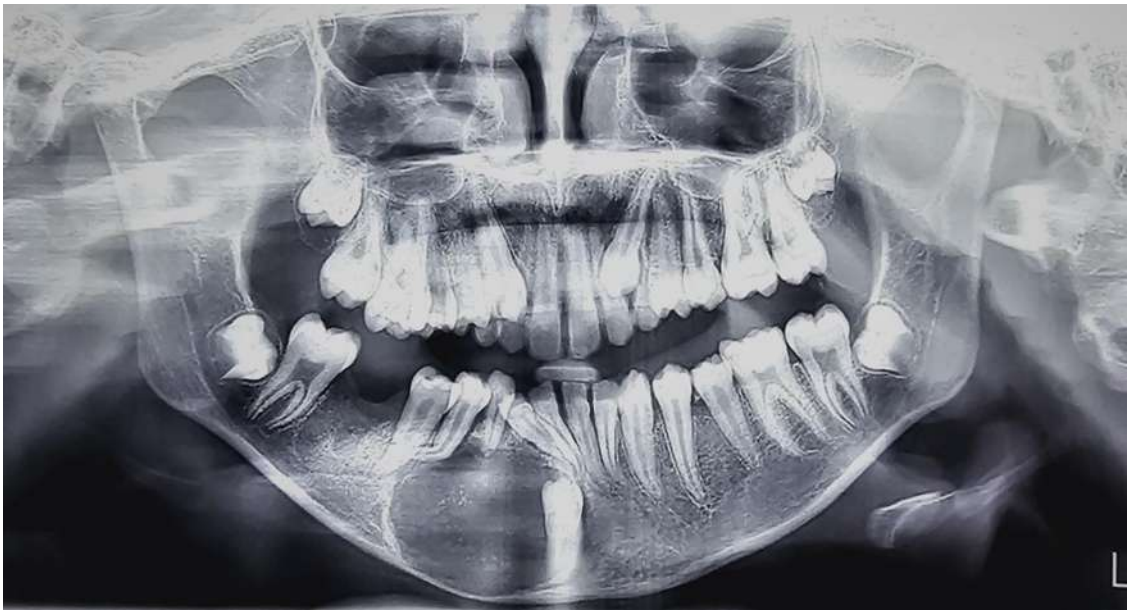
Case Report

An 11-year-old female presented with a painless swelling in the right lower anterior jaw region for 3 months, with no significant medical or family history.

Extraoral examination revealed facial asymmetry due to a diffuse, oval swelling (~3 × 4 cm) in the lower anterior mandible, extending from the chin to the premolar region and from the lower lip to the inferior border. The overlying skin was normal.with no scars, sinus, or secondary changes. On palpation, the swelling was firm, non-tender, non-fluctuant, with no local temperature rise, and the skin was freely movable. Regional lymph nodes were not palpable.

Radiographic Findings

Orthopantomogram shows a well-defined unilocular radiolucent lesion with a corticated border in the mandibular canine–premolar region, associated with an impacted 43. It causes displacement of 41 and 42 without root resorption. Internal radiopaque flecks give a "snowflake" appearance, with cortical expansion, thinning, and focal perforation, while the inferior border remains intact.



CBCT shows a well-defined, expansile mixed radiolucent–radiopaque lesion (~3.02 × 2.53 × 2.49 cm) in the right parasymphysis, associated with an impacted 43 and internal radiopaque flecks. It extends from 41 to 45, crossing the midline. Features include buccolingual expansion with cortical thinning and perforation, inferior cortical destruction, displacement of 41 and 42, root resorption of 83, loss of lamina dura in adjacent teeth, and dilaceration of 44 with cortical breach near the 46 site.

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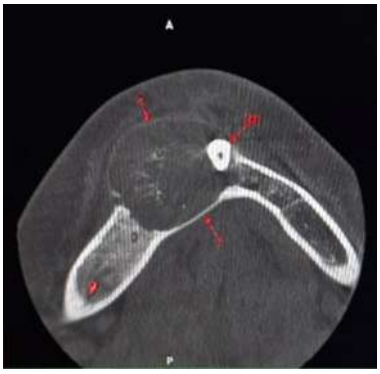
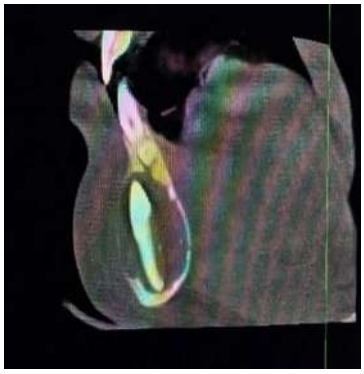
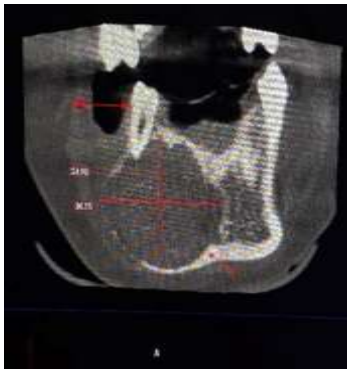
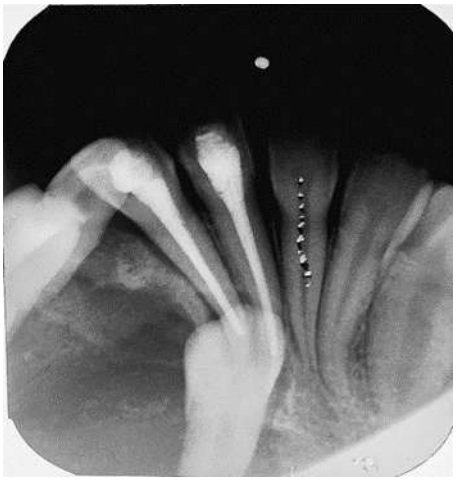
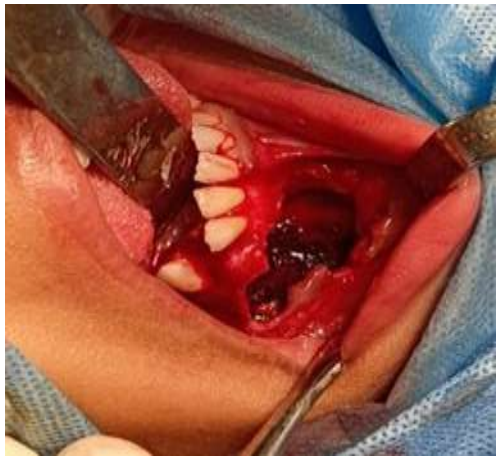




Provisional Diagnosis Dentigerous cyst Differential diagnoses Unicystic ameloblastoma, Odontogenic keratocyst, and Calcifying odontogenic cyst

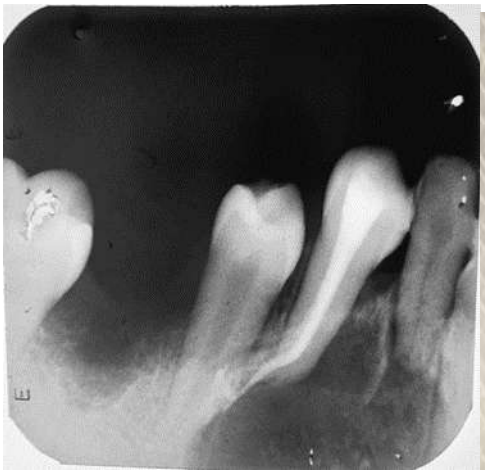
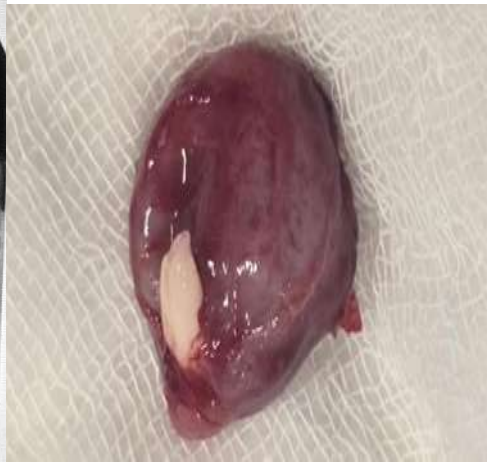
Treatment

RCT was performed for teeth 41, 42, and 44 due to periapical involvement prior to surgery. Conservative enucleation is the treatment of choice, with guided tissue regeneration considered for associated intrabony defects5.

Enucleated Site

Enucleated Lesion

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The lesion was enucleated under general anesthesia along with the impacted canine and retained 83. It was well-encapsulated, allowing complete removal. Hemostasis was achieved, Surgicel® was placed, and the site was closed with resorbable sutures. The specimen was sent for histopathological analysis. Recurrence is extremely rare, with only a few reported cases6.

Histopathological Findings:

Gross Specimen

Figure 1

Figure 2

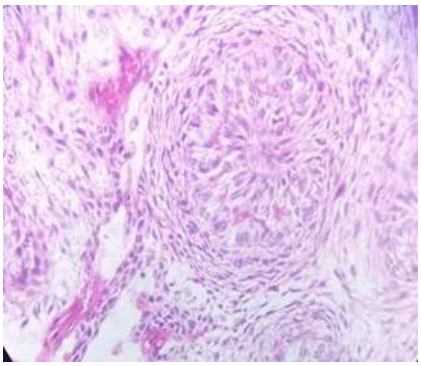
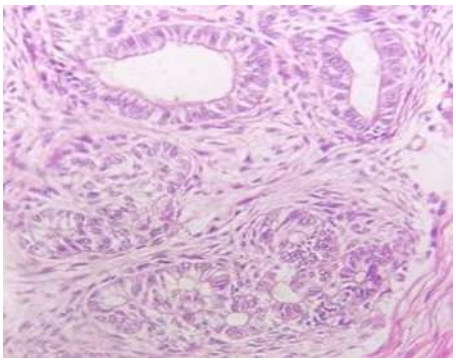


Figure 3



Sheets of polyhedral, dark-staining odontogenic epithelial cells with characteristic duct-like structures. Rosette formations with central eosinophilic material and numerous calcifications were observed. The lesion was well encapsulated by mature collagen fibers, with no evidence of atypia or malignancy, consistent with AOT

Follow-Up

Healing was uneventful, with no recurrence at 6-month follow-up.

Discussion

AOT mainly occurs in the second decade and is rare beyond 30 years. It shows a female predilection (2:1), more pronounced in Asian populations, particularly in Sri Lanka (3.2:1) and Japan (3:1). The anterior maxilla is the most common site, occurring nearly twice as often as in the mandible.2 . AOT is known as the "tumor of two-thirds," commonly occurring in the maxilla, in young females, and with unerupted canines; however, this case is atypical, involving an impacted central incisor in a male child.3. The present case deviates from the typical presentation due to its occurrence in the anterior mandible.

Epidemiology:

Since the early 1990s, 65 single cases of AOT have been reported. The mean age is 13.2 years (range: 3–28 years), with a female predominance (2.3:1). The maxilla is more commonly affected than the mandible (2.6:1). AOT accounts for about 1.2% of odontogenic tumors in Caucasians and up to 9% in black African populations. It most frequently occurs in the second decade, with females affected nearly twice as often as males4.

According to Philipsen and Reichart (1999), AOT has three variants: follicular, extrafollicular, and peripheral. The intraosseous forms (follicular and extrafollicular) account for about 96% of cases, with the follicular type being most common (71%). The follicular variant presents as a well-defined radiolucency associated with an unerupted tooth, often resembling a dentigerous cyst, whereas the extrafollicular type occurs independent of unerupted teeth and appears between or around the roots of erupted teeth, mimicking other odontogenic cysts2.

Recent studies suggest that AOT originates from the dental lamina or its remnants. The presence of CEOT-like areas within typical AOT reflects its histomorphological spectrum. Immunohistochemical and ultrastructural evidence indicates that the eosinophilic "tumor droplets" likely represent enamel matrix material.7.

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Recent studies show that about 70% of sporadic AOTs harbor KRAS mutations, though their significance remains unclear. AOT may also be associated with dental anomalies and other jaw lesions. Due to its resemblance to multiple odontogenic conditions, it is often called the "master of disguise."8.

Radiographically, AOT can mimic several odontogenic lesions, including dentigerous cysts, calcifying odontogenic lesions, ameloblastoma, odontogenic keratocyst, and periapical pathology. MRI may aid in differentiating AOT when conventional radiographs are inconclusive9.

AOT appears as a well-defined radiolucency associated with the crown of an unerupted tooth, typically up to the enamel–dentin junction. It predominantly occurs in the anterior maxilla, rarely in the mandible, and most commonly involves canines.10.Conservative surgical enucleation is the treatment of choice, and recurrence is extremely rare.The rarity of mandibular AOT highlights the importance of considering this entity in the differential diagnosis of anterior mandibular lesions in pediatric patients.

Conclusion

AOT typically occurs in the anterior maxilla, its presence in the anterior mandible is rare and diagnostically challenging, requiring histopathological confirmation. This case highlights the importance of recognizing such atypical presentations, as early diagnosis and conservative management ensure an excellent prognosis with minimal recurrence.

Acknowledgement:

We sincerely thank the Department of Paediatric & Preventive Dentistry and the Department of Oral and Maxillofacial Surgery, Government Dental College, Thiruvananthapuram, for their support.

Acknowledgements And Disclosure Statements

The authors report no conflict of interest related to this report. The authors did not receive support from any organization for the submitted work. The author-submitted authorship declaration from indicating individual contribution is available with the editorial office.

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